

Paraovarian Cysts of Neoplastic Origin Are Underreported

Noam Smorgick, MD, MSc, Arie Herman, MD, David Schneider, MD,
Reuiv Halperin, MD, PhD, Moty Pansky, MD

ABSTRACT

Introduction: We suspected that paraovarian cysts of neoplastic origin may be underreported. This study was designed to evaluate our data on the pathologic characteristics of cystic lesions located in the paraovarian area and compare them with previous studies that claimed the vast majority of these lesions were simple paraovarian cysts and only few (1.69% to 5%) were neoplastic ones.

Methods: This is a retrospective analysis of the clinical, surgical, ultrasonographic, and pathologic features of 59 women operated on for cystic paraovarian lesions at our institution from January 2002 to April 2006.

Results: Forty-four women (74.6%) had simple paraovarian cysts, and 15 (25.4%) had benign neoplastic paraovarian cysts (7 cystadenomas and 8 cystadenofibromas). There were no cases of malignant tumor. There was no difference in the clinical presentation of the women with either type of cyst. Preoperative ultrasound examinations (n=50) demonstrated more complex cysts with internal papillary projections in the group with neoplastic paraovarian cysts (41.7% compared with 7.9%, P=0.01). The macroscopic pathologic examinations revealed a significantly increased percentage of gross papillary excrescences in the group of neoplastic paraovarian cysts (10/15, 66.7%) compared with the group with simple paraovarian cysts (3/44, 6.8%, P<0.01). Other pathologic features did not differ between the 2 study groups.

Discussion: Our analysis revealed a higher percentage of paraovarian cysts of neoplastic origin (~25%) than the figures quoted in most previous reports.

Conclusion: Intraoperative inspection for diagnosing the cyst type and more frequent use of endobag devices to avoid spillage of cystic fluid are recommended.

Key Words: Paraovarian cyst, Neoplastic origin.

INTRODUCTION

Paraovarian cysts are found in the broad ligament between the ovary and the fallopian tube.^{1,2} They may be either nonneoplastic simple cysts or cysts of neoplastic origin.^{1,3} The simple paraovarian cysts originate from the embryologic remnants of the urogenital system (ie, the mesonephric and paramesonephric ducts), or from the invagination of the tube's serosa (creating a mesothelial cyst). The neoplastic paraovarian cysts originate from a neoplastic transformation of a paraovarian simple cyst or from the adjacent ovary.³ In both cases, neoplastic paraovarian cysts are generally benign serous cysts similar to benign ovarian tumors (ie, cystadenomas or cystadenofibromas),³ while borderline or even malignant paraovarian tumors are encountered less often.⁴

The prevalence of neoplastic lesions from all types of paraovarian lesions has previously been reported to be low: Genadry et al¹ found that 8 of the 140 paraovarian cysts they studied had been neoplasms (4 benign cystadenomas and 4 malignant cystadenocarcinomas, 5%), Pepe et al⁵ found only one case of benign cystadenoma in a series of 59 paraovarian cysts (1.7%), and Stein et al⁶ established that the overall incidence of malignancy in paraovarian tumors is 2%. More recently, Savelli et al⁷ found a higher prevalence of neoplastic paraovarian cysts (15 out of 50 cases, 30%).

Laparoscopy is currently the most common surgical approach in the management of paraovarian cysts. It is customary for many surgeons to either aspirate the cystic fluid via the laparoscope or to perform a fenestration of the cyst before removing it. Both techniques may cause spillage of a neoplastic cyst, with intraperitoneal dissemination of any existing malignant cells. Consequently, it is important to differentiate the simple paraovarian cysts from the neoplastic paraovarian ones in the preoperative and intraoperative settings. If a neoplastic paraovarian cyst is suspected either pre- or intraoperatively, it may be removed through an endobag, and spillage of the cyst's fluid may be avoided. Current data on the discrete char-

Department of Obstetrics and Gynecology, Assaf Harofe Medical Center, Zerifin, affiliated with the Sackler Faculty of Medicine, Tel-Aviv University, Tel-Aviv, Israel (all authors).

We thank Esther Eshkol for editorial assistance.

Address correspondence to: Moty Pansky, MD, Department of Obstetrics and Gynecology; Assaf Harofe Medical Center, Zerifin, 70300 Israel. Telephone: 972-8-9779000-1, Fax: 972-8-9778026, E-mail: motyp@asaf.health.gov.il

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acteristics of simple versus neoplastic paraovarian cysts are limited. Previous studies did observe that neoplastic paraovarian cysts usually involved cysts measuring $>5\text{cm}$,⁶ but other clinical or surgical differentiating criteria were not investigated.

A similar clinical dilemma exists with regard to the management of ovarian cysts. In cases of ovarian cysts, however, a preoperative ultrasound scan is usually performed, and its results may be helpful in distinguishing between benign and malignant cysts.⁸ Fewer data are available on the ultrasound features of the paraovarian simple compared with neoplastic cysts. In the few series that described the ultrasound characteristics of the 2 paraovarian cyst types,^{7,9-11} simple paraovarian cysts were usually visualized as simple unilocular cysts while paraovarian cystadenomas were usually described as complex cysts having a nodular solid component.^{7,9}

The current study is a retrospective review of the clinical, sonographic, and pathological characteristics of intraoperatively identified paraovarian cystic masses. The different features of simple paraovarian cysts were compared with those of neoplastic paraovarian cysts in an attempt to establish distinguishing criteria between them.

MATERIALS AND METHODS

Fifty-nine women with an operative diagnosis of paraovarian cyst(s) were identified through a computerized search of our laparoscopic surgery registry (January 2002 through April 2006), and their medical records were reviewed. All the women were managed by laparoscopic surgery (either cystectomy or adnexectomy). The reasons for surgery were a pelvic mass incidentally found by sonography, part of an abdominal pain workup, or a suspected adnexal torsion or ectopic pregnancy. The surgical reports were reviewed for the description of the cysts' location, as well as for their size, the number of loculations, and the possible torsion of the adnexa. The final analysis included the cysts that were described at a paraovarian location and whose removal did not involve a dissection of the ovarian capsule (ie, the cysts that were clearly paraovarian as opposed to ovarian). The pathologic examination included a macroscopic specimen evaluation consisting of the cyst size, loculations, the presence of papillations on external or internal cyst walls, and the wall thickness, as well as a microscopic examination. The paraovarian masses were divided into simple paraovarian cysts (ie, of paramesonephric, mesonephric, and mesothelial origin) versus neoplastic paraovarian cysts, according to the final pathologic findings. All the neoplastic

cysts in the current study were benign serous cystadenomas or benign serous cystadenofibromas. There were no cases of malignancy.

Additional information, retrieved retrospectively from the medical records of the 59 women, included the clinical presentation and the menopausal status (available for all subjects), a preoperative pelvic sonogram (available for 50 women), and a preoperative CA-125 level (available for 24 women).

Statistical analysis was performed using Win-Pepi software. The Student *t* test and the Fisher exact test were used as appropriate. A *P* value <0.05 was considered statistically significant.

The institutional review board approved the study.

RESULTS

Fifty-nine women with adnexal masses located in the paraovarian area were included in the study. Forty-four cysts (74.6%) were of the simple paraovarian type and 15 (25.6%) were benign neoplastic cysts. Cystectomy was performed in 43 cases (72.9%) and adnexectomy in 16 (27.1%). The reasons for adnexectomy were age >45 years (in all cases of adnexectomy) combined with either history of breast carcinoma (in 6.3% of adnexectomy cases), elevated CA-125 levels (12.5%), or preoperative ultrasound scan suspicious for malignancy (56.2%). Torsion of the ipsilateral adnexa involving the paraovarian mass was found in six women (10.2%), and they underwent detorsion and cystectomy. **Table 1** lists the clinical characteristics of the women with simple paraovarian cysts and those with paraovarian cystadenoma/cystadenofibroma, as well as the intraoperative appearance of the cyst. There was no difference in the age or menopausal status between the women in the two groups. The mean size of the cyst was similar in both groups ($6.4 \pm 3.4\text{cm}$ and $6.3 \pm 2.9\text{cm}$, respectively, $P=0.9$). Simple paraovarian cysts were distributed almost equally on the right and left side, but paraovarian cystadenoma/cystadenofibroma were more prevalent on the left side (60%, $P=0.5$). Indications for surgery were similar in both groups (**Table 1**).

Preoperative ultrasound examination was available in 50 cases (**Table 2**). The most common ultrasonic presentation of simple paraovarian cysts was a simple uniloculated cyst (84.2%). Complex cysts with internal papillary projections were significantly more frequent in paraovarian cystadenomas/cystadenofibromas than in simple paraovarian cysts (41.7% vs. 7.9% respectively, $P=0.01$). The

Table 1.
Clinical and Surgical Characteristics of the 59 Studied Women

| Patient Characteristics | Simple Paraovarian Cysts (N = 44) | Neoplastic Paraovarian Cysts (N = 15) | P Value |
|---------------------------|--------------------------------------|--|---------|
| Age (years) | 33 ± 14.4 (range, 16 to 70) | 36.6 ± 16.8 (range, 17 to 70) | 0.4 |
| Menopausal Status (%) | | | |
| Premenopausal | 38 (86.4) | 10 (66.7) | 0.1 |
| Postmenopausal | 6 (13.6) | 5 (33.3) | 0.1 |
| Cyst Size (cm) | 6.4 ± 3.4 | 6.3 ± 2.9 | 0.9 |
| Location (%) | | | |
| Right | 23 (52.3) | 5 (33.3) | 0.2 |
| Left | 21 (47.7) | 9 (60) | 0.5 |
| Bilateral | 0 | 1 (6.7) | 0.2 |
| Clinical Presentation (%) | | | |
| Ultrasonic finding | 23 (52.3) | 9 (60) | 0.8 |
| Abdominal pain workup | 7 (15.9) | 4 (26.7) | 0.4 |
| Torsion of adnexa | 5 (11.3) | 1 (6.6) | 1.0 |
| Acute abdominal pain | 8 (18.2) | 1 (6.6) | 0.4 |
| Ectopic pregnancy | 1 (2.3) | 0 | 1.0 |

Table 2.
Ultrasound Features of the 50 Scanned Cysts

| | Simple Paraovarian Cysts (N = 38) | Neoplastic Paraovarian Cysts (N = 12) | P Value |
|------------------------------|--------------------------------------|--|---------|
| Uniloculated simple cyst (%) | 32 (84.2) | 7 (58.3) | 0.1 |
| Biloculated cyst (%) | 3 (7.9) | 0 | 1.0 |
| Presence of papillations (%) | 3 (7.9) | 5 (41.7) | 0.01 |

average size of the papillations was 9.75 ± 2.5 mm in the paraovarian cystadenomas/cystadenofibromas.

Preoperative CA-125 levels were determined in 24 women and found to be elevated in 2 (8.3%). It was 103 U/mL (normal = <35 U/mL of serum) in a 53-year-old woman who was found at laparoscopy to have a right paraovarian cyst and a left endometrioma. The elevated CA-125 was attributed to endometriosis. In the second case, the preoperative CA-125 was 146 U/mL in a 44-year-old woman in whom a left paraovarian cyst was found at laparoscopy. The second patient was a premenopausal, otherwise healthy woman, and the adnexal cystic mass was discovered on a routine sonogram. We could not account for her elevated CA-125 level.

The macroscopic pathologic characteristics are listed in **Table 3**. All neoplastic cysts were of the benign serous

type, including 7 serous cystadenomas and 8 serous cystadenofibromas. Most cysts in the current study were uniloculated cysts (95.4% of simple paraovarian cysts and 86.7% of neoplastic paraovarian cysts, $P=0.3$). While all the paraovarian cysts (of both types) had smooth, thin external walls, gross papillary excrescences were significantly more prevalent in the neoplastic than in the simple paraovarian ones (66.7% and 6.8%, $P<0.01$).

DISCUSSION

Four previous studies^{1,5-7} have analyzed the incidence of neoplastic paraovarian cysts from all their reported paraovarian cystic lesions. The first 3 studies^{1,5,6} reported a combined very low incidence of neoplastic paraovarian cysts (12 neoplastic cysts from among 367 paraovarian cystic lesions, 3.3%). This discrepancy might be explained

Table 3.
Pathologic Features of the 59 Studied Cysts

| | Simple Paraovarian Cysts (N = 44) | Neoplastic Paraovarian Cysts (N = 15) | P Value |
|---|--------------------------------------|--|---------|
| Cyst Loculations (%) | | | |
| Uniloculated | 42 (95.4) | 13 (86.7) | 0.3 |
| Biloculated | 1 (2.3) | 0 | 1.0 |
| Multiple loculations | 1 (2.3) | 2 (13.3) | 0.1 |
| Cyst Walls (%) | | | |
| Smooth external walls | 44 (100) | 15 (100) | |
| Presence of internal papillary excrescences | 3 (6.8) | 10 (66.7) | <0.01 |

by the different methodologies used in those studies compared with ours. Specifically, the reports of Genardy et al,¹ Pepe et al,⁵ and Stein et al⁶ were pathologic series, meaning that the authors retrospectively identified the relevant cases by searching the records of their respective pathology departments for the diagnosis of paraovarian cysts. Our current study is primarily a series of surgical cases identified by examining operative reports, and it includes those cysts that were intraoperatively diagnosed at a paraovarian location, the only point where pathologic diagnosis was correlated. The different approaches of the previous studies versus the current study may explain the higher rate of neoplastic cysts in the latter. Since the microscopic appearance of paraovarian cystadenomas and ovarian cystadenomas are indistinguishable, the pathologist depends entirely upon correct reporting of the cyst's location. In the event of incorrect reporting, a paraovarian cystadenoma may be mistaken for an ovarian cystadenoma, ultimately leading to an underestimation of the former's occurrence. A fourth study by Savelli et al⁷ more recently described a higher prevalence of neoplastic paraovarian cyst reaching 30%. These results are similar to the findings of the current study.

We found that both simple and neoplastic paraovarian cysts occurred over a wide age range, including adolescents, women of reproductive age, and postmenopausal women. There was no statistically significant difference in age, menopausal status, or clinical presentation between the 2 groups. At laparoscopy, both types of cysts appeared as smooth, thin-walled masses located in the broad ligament. Although there was a great variation in size, the mean cyst diameter was not statistically different between the 2 groups and could not be used as a differentiating criterion. As for the internal cystic wall, we did find that gross papillary excrescences characterized the neoplastic

cysts and not the simple cysts, as was previously reported by others.³

The ultrasonographic features of simple and neoplastic paraovarian cysts were described previously in several series.^{7,9-11} Kim et al⁹ and Alpern et al¹⁰ demonstrated most of the paraovarian cysts as simple unilocular cysts. Korbin et al¹¹ described the sonographic features of 14 paraovarian cystadenomas: 3 of them appeared as simple cysts, while 9 appeared as complex cysts with a nodular solid component, and the remaining as septated cysts. Savelli et al⁷ described the histology of 15 paraovarian cysts containing papillary projections on preoperative ultrasound. They found 8 cystadenofibromas, 5 cystadenomas, and 2 serous papillary borderline tumors. Similarly, we compared the ultrasound features of simple and neoplastic paraovarian cysts: the simple ones were visualized as simple unilocular cysts in 32/38 (84.2%) cases, while neoplastic paraovarian cysts were visualized as simple cysts in 7/12 (58.3%) cases and as complex cysts with a major cystic component and small papillary projections (<1 cm) in 5/12 (41.7%) cases. This difference was statistically significant (P<0.01).

The consequences of intraoperative spillage of paraovarian cysts are to date unknown. However, a similar consideration concerns the removal of ovarian cysts suspicious for malignancy. In the latter case, intraoperative spillage with dissemination of malignant cells in the peritoneal cavity has been shown to reduce patients' 5-year survival.¹² Thus, intraoperative spillage of suspicious ovarian cysts should be avoided. Since a considerable number of neoplastic paraovarian cysts were found in our series, a more prudent approach in the management of paraovarian cysts would be to avoid spillage by using endoscopic retrieval bags.

CONCLUSION

We found a higher percentage of paraovarian cysts of neoplastic origin (~25%) than previously reported. Preoperative information, such as patient's age, menopausal status, and clinical presentation, cannot be used for differential diagnosis between simple and neoplastic paraovarian cysts, whereas preoperative ultrasonography showing papillary projections and gross papillary excrescences observed on the internal cysts' walls may point to neoplastic paraovarian cysts. Intraoperative inspection for diagnosing the cyst type, frequent use of endobag devices to avoid spillage, and judicious use of frozen section examination are recommended. This recommendation is supported by the considerable percentage of malignant neoplastic paraovarian cysts reported in previous series.⁶

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